CASE REPORT

WILEY

Clinical Case Reports

Lymphedema: An early sign of rodent ulcer metastasis leads to timely intervention

Caroline Mann¹ | Stephan Rietz¹ | Ronald Amedee² | Hadrian Schepler¹

Correspondence

Caroline Mann, Department of Dermatology, University Medical Center, Langenbeckstr. 1, 55131 Mainz, Germany. Email: doc.caroline.mann@gmail.com

Abstract

Basal cell carcinoma has a potential for early and late metastasis. Depending on the location of the primary site, the relevant lymphatic drainage routes have to be monitored. Regional lymph edema may be a first indicator for metastasizing disease.

KEYWORDS

basalcell carcinoma metastasis, lymphedema, ultrasound, surgical intervention

1 | INTRODUCTION

Basal cell carcinoma is the most common skin cancer developing with various clinical and histologic features mostly in parts of the body exposed to UV-light. Especially, the head and neck area is at risk. Some tumors develop in other parts of the body and may be part of a rare genetic syndrome. ¹

Therapy for locally advanced tumors includes surgical excisions with clear margins, laser or radiotherapy, and in advanced stages hedgehog pathway inhibitors, chemotherapy, and PDL-1 immunotherapy in study protocols.² Metastatic disease is unexpected and occurs according to the literature in only 0.025%-0.55%.³

2 | CASE REPORT

A 50-year-old otherwise healthy, nonimmunosuppressed male presented with several basal cell carcinomas and a rodent ulcer, on his right shoulder, which was resected with clear margins followed by radiotherapy to the primary tumor site (Figure 1). Because of the size of the primary tumor, 60 Gy was administered.

Initial ultrasound examination of the right axilla showed a lymph node measuring $1.8 \times 1 \times 1$ cm, which was classified as unspecific lymph node enlargement. The patient presented

3 years after initial treatment with sudden extensive lymph edema of the right upper extremity. On clinical examination, there was no lymph node enlargement palpable in the right axilla; however, ultrasound revealed only one suspicious lymph node (Figure 2).

Preoperative PET-CT demonstrated two lymph nodes in the right axillary region and no evidence of further metastasis (Figure 3). The patient was admitted for lymphadenectomy. Histologic examination revealed only one metastatic lymph node with basaloid differentiation and perinodular infiltration and one lymph node with unspecific inflammation.



FIGURE 1 Rodent ulcer of the right shoulder before surgical intervention

This is an open access article under the terms of the Creative Commons Attribution-NonCommercial-NoDerivs License, which permits use and distribution in any medium, provided the original work is properly cited, the use is non-commercial and no modifications or adaptations are made.

© 2019 The Authors. Clinical Case Reports published by John Wiley & Sons Ltd.

Clin Case Rep. 2019;7:1551–1553. wileyonlinelibrary.com/journal/ccr3

¹Department of Dermatology, Johannes Gutenberg University, Mainz, Germany ²Department of ENT, Ochsner Medical Center New Orleans, New Orleans, Louisiana, USA

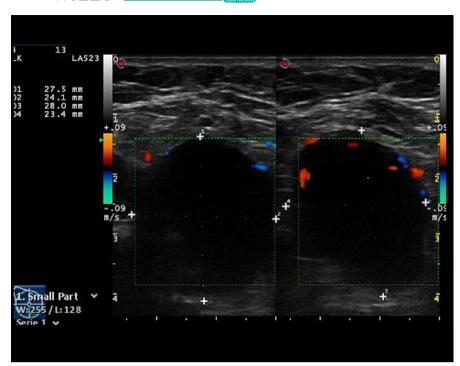


FIGURE 2 Hypoechogenic, roundshaped lymph node in the right axillary region showing a loss of hilum with peripheral perfusion



FIGURE 3 PET-CT scan showing lymphedema of the right arm and localized FDG avidity in the right axilla

In addition, final histopathology revealed carcinomatous hemangiosis. Because of previous radiotherapy, only limited additional radiotherapy with 50 Gy was feasible. After 12 months, there is no sign of recurrent disease.

3 | DISCUSSION

This is the first time that lymphedema of the upper extremity was indicative for metastatic disease in basal cell carcinoma. Since his first description in 1894 by Beadles et al, only approximately 400 cases of metastatic disease have been reported in the literature. ^{4,5} It remains to be debated if the axilla lymph node at the time of primary treatment was involved by micrometastasis and included in the field of radiotherapy with later growth leading to lymphedema.

4 | CONCLUSION

There are still ongoing discussions how to treat metastasized basal cell carcinoma, thus making individual strategies necessary.

Individualized therapy depends on tumor load and localization. Today's options include hedgehog inhibitors, surgery, and radiotherapy. Recent studies include PD1 inhibitor therapy and tumor vaccination (Jäger, unpublished data/personal communication). In this case, lymphedema, as an early warning sign, led to timely intervention and further recurrence free survival till date.

CONFLICT OF- INTEREST

None declared.

AUTHOR CONTRIBUTIONS

CM: is a patient physician and main clinical researcher. SR: is a consultant reviewing the clinical data. RA: is a senior advisor and academic teacher. HS: is a senior author and primary surgeon.

ORCID

Caroline Mann https://orcid.org/0000-0001-7044-2216

REFERENCES

- 1. Lo ML. Nevoid basal cell carcinoma syndrome (Gorlin syndrome). *Orphanet J Rare Dis.* 2008;3:32.
- 2. Fischer S, Ali OH, Jochum W, Kluckert T, Flatz L, Siano M. Anti-PD-1 therapy leads to near-complete remission in a patient with metastatic basal cell carcinoma. *Oncol Res Treat*. 2018;41(6):391-394.
- 3. Piva de Freitas P, Senna CG, Tabai M, Chone CT, Altemani A. Metastatic basal cell carcinoma: a rare manifestation of a common disease. *Case Rep Med.* 2017;2017:8929745.

- 4. Beadles CF. Rodent ulcer. Tran Pathol Soc Land. 1894;45:176-181.
- Rappoport WD. Metastatic basal cell carcinoma. Otorrinolaringol Cir Cabeza Cuello. 2016;76:347-352.

How to cite this article: Mann C, Rietz S, Amedee R, Schepler H. Lymphedema: An early sign of rodent ulcer metastasis leads to timely intervention. *Clin Case Rep.* 2019;7:1551–1553. https://doi.org/10.1002/ccr3.2285